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Idiopathic choroidal neovascular membrane in a child

Membrana neovascular subretiniana idiopática em uma criança

Membrana neovascular subretiniana idiopática en un niño

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ABSTRACT

A case of idiopathic choroidal neovascular membrane (CNVM) is described in a 12-year-old male patient. On initial examination, his vision was 20/100 in the left eye (OS). Fluorescein angiography and optical coherence tomography scan (OCT) showed a classic type 2 choroidal neovascular membrane. Intravitreal ranibizumab was injected into the OS, three times, with an interval of 30 days between injections. At a follow-up visit, 3 weeks after the third injection, his best corrected visual acuity in the left eye was 20/20, with complete resolution of fluid on the OCT. This case is unusual, in that the CNVM developed in a young patient with no significant past medical history and in the absence of a choroidal or retinal pigment epithelial disease process that may be associated with a CNVM.

RESUMO

Um caso de membrana neovascular subretiniana idiopática (CNVM) é descrito em um paciente de 12 anos, do sexo masculino. No exame inicial, sua visão era de 20/100 no olho esquerdo (OE). A angiografia por fluoresceína e a tomografia por coerência óptica (OCT) mostraram uma membrana neovascular coroidal clássica do tipo 2. O ranibizumab intravítreo foi injetado no OE, três vezes, com um intervalo de 30 dias entre as injeções. Em uma visita de acompanhamento, 3 semanas após a terceira injeção, sua acuidade visual melhor corrigida no olho esquerdo foi de 20/20, com resolução completa de fluido na OCT. Este caso é incomum, na medida em que a CNVM se desenvolveu em um paciente jovem sem história médica passada significativa e na ausência de um processo epitelial de pigmento coroideo ou retiniano que poderia estar associado a uma CNVM.

RESUMEN

Un caso de membrana neovascular subretiniana (MNVS) idiopática es descrito en un paciente de 12 años de edad, del sexo masculino. En el análisis inicial, su mejor agudeza visual corregida era 20/100 en el ojo izquierdo. La angiografía fluoresceínica y Tomografía de Coherencia Óptica (TCO) mostraron una MNVS clásica, tipo 2. Inyecciones intravítreas de Ranibizumab se realizaron tres veces en el ojo izquierdo, con un intervalo de 30 días entre ellas. En el regreso, tres semanas después de la tercera inyección, el paciente presentaba mejor agudeza visual TCO de 20/20 en el ojo izquierdo, con completa resolución de los fluidos en el OCT. Este caso es infrecuente, considerando que la MNVS se desarrolló en un paciente joven, sin historial médico significativo y ausencia de procesos patológicos de la coroides o del epitelio pigmentar de la retina, que generalmente se encuentran asociados a la MNVS.

Keywords:
Choroidal Neovascularization;
child;
Ranibizumab.

Palavras-Chave:
Neovascularização de Coroide;
criança;
Ranibizumab.

Palabras Clave:
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INTRODUÇÃO

Choroidal neovascularization (CNV) is characterized by the growth of new blood vessels that originate from the choroid through a break in Bruch's membrane into the sub-retinal pigment epithelium (sub-RPE) or subretinal space[1,2]. The neovascular membrane usually occurs at the macula or at the margin of the optic disc and often leaks blood and fluid, resulting ultimately in photoreceptor cell death [1].

The most common cause of CNV in adults is age-related macular degeneration, the leading cause of blindness in Europe and North America [1,2]. Subfoveal choroidal neovascularization in children is a rare event, typically occurring as a complication of inflammatory or infectious chorioretinal disease [3].

Other causes of pediatric CNV, including developmental abnormalities, dystrophies, and trauma, have been described [4,5,6]. However, in a significant number of young patients with CNV, no apparent cause can be detected, constituting idiopathic CNV [7,8]. These membranes are usually unilateral and final visual outcomes are considered to be more favorable than those of CNV due to age-related macular degeneration (AMD) [8,9]. Only a few reports exist in the literature describing idiopathic CNV in young patients [8,10]. Current management options for pediatric subfoveal CNV include observation, laser photocoagulation, photodynamic therapy, anti-vascular endothelial growth factor therapy (VEGF), and submacular surgery [6,11].

In this report, we describe the case of a 12-year-old boy diagnosed with idiopathic choroidal neovascular membrane in the left eye, successfully treated with three doses of intravitreal ranibizumab.

CASE REPORT

A 12-year-old male student was referred for evaluation of decreased vision in his left eye (OS). There was no history of pain, redness, or photophobia. There was no history of trauma. Past medical history was unremarkable. On examination, his best corrected visual acuity was 20/20 in the right eye (OD) and 20/100 in the left eye (OS). His anterior segment examination was normal in both eyes (OU). On biomicroscopy of the posterior pole of the left eye, subfoveal choroidal neovascularization with subretinal hemorrhage was noted. Fundus examination of the right eye was unremarkable. No drusen, retinal pigment epithelial changes, or macular exudates were observed OU.

Fluorescein angiography (Figure 1) showed a well-defined hyperfluorescent area corresponding to the CNV on the left eye with diffuse leakage in the late phase. An optical coherence tomography scan (OCT) showed a classic type 2 choroidal neovascular membrane (Figure 2). After informed consent was obtained, the patient was sedated with Propofol and the left eye was anesthetized using topical tetracaine 1% drops and then intravitreal ranibizumab (Lucentis) was injected on the left eye supratemporally 3.5 mm posterior to the limbus at a dose of 0.05 ml/0.5 mg. This procedure was repeated three times with an interval of 30 days between injections. At a follow-up visit, 3 weeks after the third injection, his best corrected visual acuity in the left eye was 20/20, with complete resolution of fluid in the OCT (Figure 3). The patient remains under follow-up and his last OCT, carried out 5 months after the last injection, demonstrated the absence of fluid with scarred choroidal neovascular membrane in the parafoveal region (Figure 4).

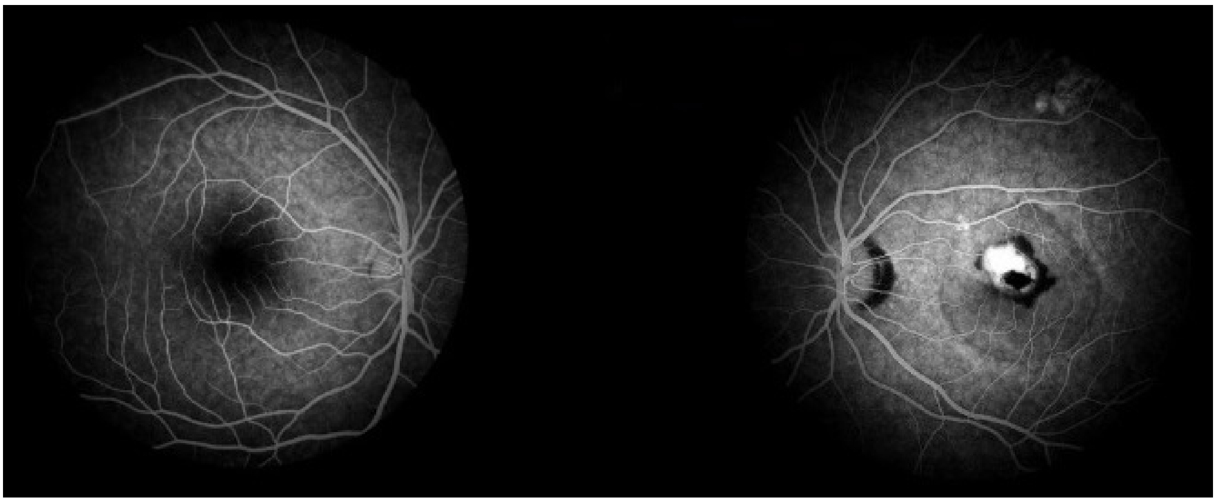


Figure 1

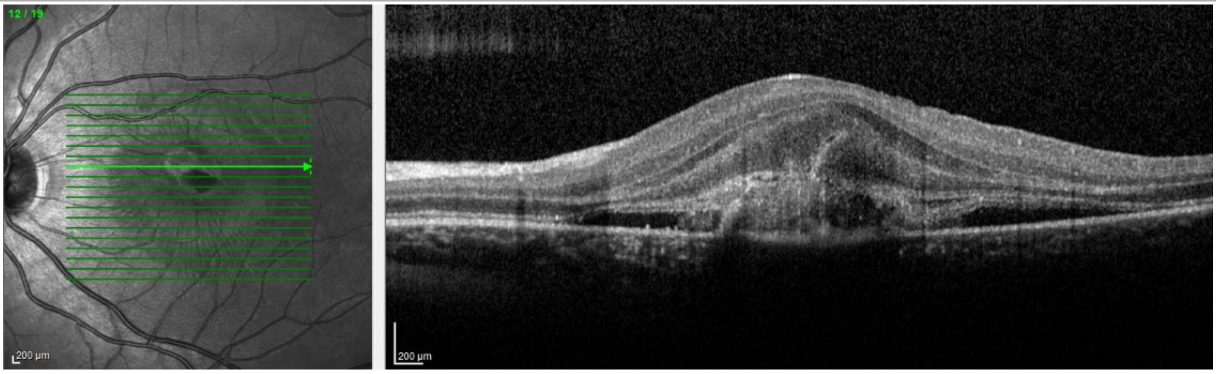


Figure 2



Figure 3

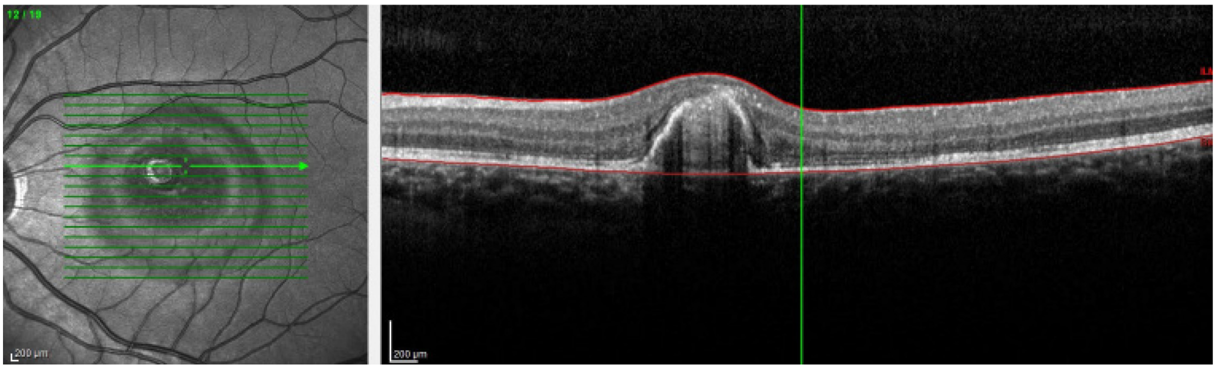


Figure 4

DISCUSSION

Although the clinical course of subfoveal CNV secondary to AMD is well documented in the literature, sparse information exists on both the natural history and treatment of CNV in the pediatric age group. Due to the rare occurrence of this entity, there are not many published reports in the available literature, which sometimes poses some challenges in the management of this condition[3].

Different treatment approaches for patients with CNV have been described: thermal laser therapy, ocular photodynamic therapy with verteporfin, transpupillary thermotherapy, submacular surgery, intravitreal injections of anti-VEGF agents[8] or observation [1].

Laser photocoagulation of extrafoveal and juxtafoveal CNV can usually be safely performed in teenagers and older cooperative children using similar techniques to those used in adults [1]. On the other hand, as the natural history of idiopathic CNV is better than that seen in AMD, thermal laser therapy does not appear to be a suitable treatment option because of the risk of immediate central vision diminution. Photodynamic therapy is too expensive and usually requires repeated sessions[8].

Von Eiken et al. reported successful photodynamic therapy in one case of a 5-year-old girl with subfoveal CNV [12]. Transpupillary thermotherapy is outdated and is widely replaced nowadays by anti-VEGF agents [8].

Previous studies of submacular surgery in pediatric CNV have reported good visual outcomes for membranes of various etiologies, with 92% of patients having an improvement in visual acuity. However, some eyes never attained vision better than 20/400, and there have been reported recurrences[6]. In a series reported by Goshorn et al., 58% of untreated patients underwent spontaneous involution, with a final visual acuity of better than 20/50. However, in their series, 90% of patients with initial visual acuity of less than 20/200 achieved final visual acuity of less than 20/80. The CNVs were of multiple causes, and 5 of the 11 CNVs were extrafoveal or peripapillary[13].

The successful use of anti-VEGF therapies in younger patients, most notably in neonates with retinopathy of prematurity, has allowed the application of this treatment to other pediatric conditions [3]. Cakir and colleagues reported two children with choroidal neovascular membrane that regressed following treatment with bevacizumab with documented improvement in visual acuity[10]. Kohly et al. presented four cases where the use of intravitreal anti-VEGF agents for the treatment of pediatric CNVs resulted in an improvement or stabilization of VA and a significant resolution of fluid on OCT[11]. Mandal et al. reported results of intravitreal bevacizumab (1.25 mg/0.05 mL) in 32 eyes with idiopathic subfoveal CNV. After 12 weeks of follow-up, 19 eyes (59%) had an improvement in BCVA of three or more lines, 11 eyes (34%) remained stable and two eyes (6%) lost three or more lines. Their observations suggest that short-term use of intravitreal bevacizumab is safe and well tolerated in the management of idiopathic CNV[5]. Bevacizumab injection should be repeated if OCT shows intraretinal/subretinal fluid and/or pigment epithelial detachment at a 4–6-week interval [8].

Although intravitreal anti-VEGF injections appear to have a low complication rate, possible ocular complications include bacterial endophthalmitis, retinal detachments, and uveitis [14]. Systemic side effects following intravitreal anti-VEGF injections have been an area of debate. Acute elevation of blood pressure and stroke have been reported [15]. All these must be made known to the patient and his or her family, and informed consent must be obtained prior to therapy [8].

CONCLUSION

Ongoing studies monitoring ocular and systemic toxicities are vital in establishing the long-term safety profile of anti-VEGF drugs in children. In our patient, the CNV regressed with complete resolution of the intraretinal fluid, visual acuity improved significantly after ranibizumab injections, and no adverse effects attributable to the drug or procedure were encountered in the follow-up period. However, our patient is in follow-up, and a new injection will be performed if there is recurrence of the CNV. We cannot comment on the efficacy, limitations, and long-term side effects of the treatment based on our case. We recommend a multicenter, prospective randomized controlled study to address the issue.

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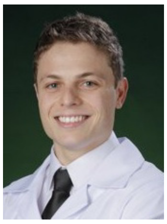
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